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Robot assisted laparoscopic partial cystectomy for inflammatory myofibroblastic tumor with simultaneous intraoperative flexible cystoscopy for tumor mapping

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ABSTRACT

Bladder masses are an infrequent occurrence rarely suspected in cases of pediatric hematuria. Inflammatory myofibroblastic tumors represent one differential diagnosis that is difficult to characterize as purely benign and should therefore be given special consideration. Although uncommon, this is an important entity to recognize for potential bladder sparing and minimally invasive surgical approaches.

1. Introduction

Inflammatory myofibroblastic tumors (IMTs) represent a rare cause of gross hematuria in the pediatric population.[1] Approximately 40–50 cases have been described in children. Case reports of metastatic and locally invasive tumors have been described in the adult population.[2] Although these tumors can sometimes respond to medical therapy, extirpative surgery remains the standard for definitive treatment.[3] We present a 6-year-old female diagnosed with IMT and managed with robot assisted partial cystectomy aided by simultaneous intraoperative flexible cystoscopy for tumor mapping.

2. Case presentation

A 6-year-old female presented to the emergency department with recurrent dysuria and hematuria one month after presentation for suspected urinary tract infection. Ultrasound was performed demonstrating 3.6cm intraluminal irregular mass. A CT scan further demonstrated a complex, infiltrative mass with cystic components protruding into the bladder lumen (Fig. 1). The patient was admitted for further workup with cystoscopy and transurethral resection (TUR) as well as exam under anesthesia. Cystoscopy revealed an intravesical bladder mass which was exophytic and lobulated. Bimanual exam revealed a 4cm round, firm mobile mass on the right side of the bladder. Diagnosis of IMT was obtained via TUR specimen.

Indications for definitive excision were discussed with the family who wished to proceed. The patient underwent robot assisted laparoscopic partial cystectomy with intraoperative flexible cystoscopy for tumor mapping. The tumor involved the detrusor near the right ureteral tunnel. A feeding tube was placed in the ureter to aid in dissection and excision of the mass. A ureteral stent with string was placed after resection given the amount of ureteral manipulation. A two-layer bladder closure was performed and a foley catheter was left in place (Fig. 2, video link). She was discharged on post-operative day 2. One-week post-surgery, cystogram showed no extravasation and the catheter and stent were removed. 6-month post-operative ultrasound showed no hydronephrosis. Final pathology confirmed IMT with negative surgical margins.

Supplementary video related to this article can be found at https://doi.org/10.1016/j.eucr.2022.102070

The right ureter is identified at its insertion into the bladder. Intraoperative cystoscopy is utilized to delineate the borders of the mass. The tumor involved the detrusor near the right ureteral tunnel. A feeding tube was placed in the ureter to aid in dissection and excision of the mass, and later exchanged for a ureteral stent with string given the amount of ureteral manipulation. The bladder was closed in two layers.

3. Discussion

IMTs most often occur in the lungs and rarely occur in the urinary

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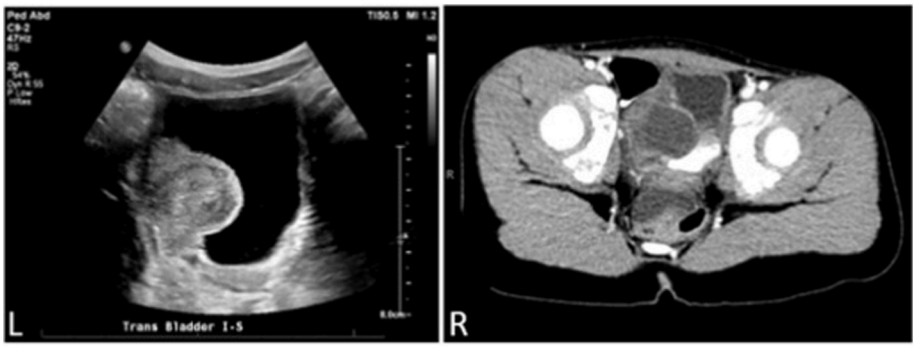

Fig. 1. Left: Transverse representative ultrasound image demonstrating intraluminal bladder mass. Right: Axial view of bladder mass on CT with oral and intravenous contrast.

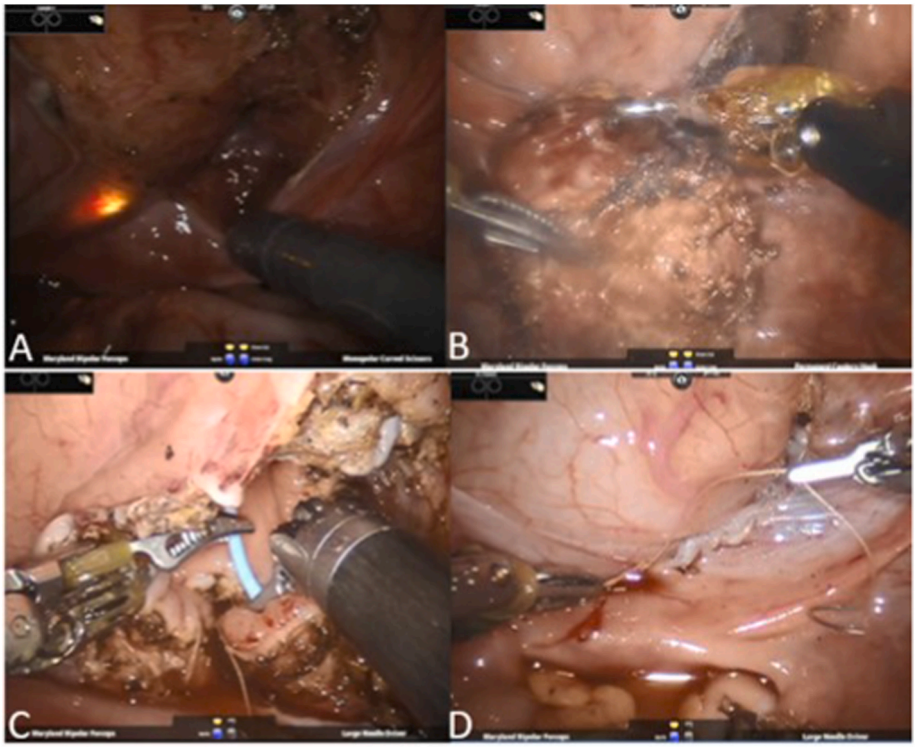

Fig. 2. A: Demonstration of normal bladder mucosa adjacent to the tumor utilizing intraoperative flexible cystoscopy with diminished luminosity at the robotic console. B: Complete excision of the tumor. C: Stent placement secondary to proximity of the mass to the right ureteral orifice and tunnel. D: two-layer bladder closure.

bladder. A subset can be associated with high-grade urothelial carcinoma, largely in the adult population.[4] Although they largely follow an indolent course, pathologic analysis with immunohistochemical staining must be utilized to differentiate them from malignant masses. The classification of these lesions as benign has largely been challenged by findings of chromosomal abnormalities, local recurrence, and rare reports of metastatic disease.[5] No recurrences have been reported in the pediatric population to our knowledge. Minimally invasive, bladder sparing approaches have been described as a management option in the adult, but less is reported about pediatric minimally invasive management. Medical therapy with cyclooxygenase-2 inhibitors may serve a role in tumor size reduction to allow for partial cystectomy.[3]

4. Conclusion

IMTs are a rare cause of gross hematuria in the pediatric population. When appropriately localized, minimally invasive bladder preserving techniques can be implemented to achieve complete surgical resection with excellent outcomes.

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